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Angioleiomyoma of the upper lip: report of a case

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Abstract. A rare case of vascular leiomyoma of the upper lip in a 51-year-old man is presented. The differential diagnosis, frequency, treatment, and prognosis are discussed.

Key words: angioleiomyoma; vascular; leiomyoma; upper lip.

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Leiomyomas are benign mesenchymal tumors derived from smooth muscle. They are subdivided into three groups: vascular (angioleiomyomas), which comprise approximately 74%; solid, which comprise 25%; and epitheloid (leiomyoblastomas), which are extremely rare (less than 1%)[2]. Angioleiomyomas are uncommon, slow-growing, dermal or subdermal tumors[9]. They are most commonly located at the anterior aspect of the lower extremities (approximately 70%)[7], and affect predominantly middle-aged women. The face is rarely affected – 5–10% of all angioleiomyomas. The predilection sites for oral leiomyomas are as follows: lip – 26%; tongue – 21%; palate – 21%; cheek – 18%; and others – 14% (gingivae, floor of the mouth, etc.)[9]. There are currently only 84 reported cases of oral angioleiomyomas, of which 15 occurred on the upper lip[9].

Case report

A 51-year-old Caucasian man was referred to the oral and maxillofacial department of the University Hospital of Nijmegen, The Netherlands, by his family physician for removal of a "mucocele" of the upper lip. He had a small, painless swelling, measuring approximately 1.5×1 cm, with a reddish/bluish overlying mucosa, present for about 1 year (Fig. 1). The past medical history was unremarkable. The tumor appeared to be mobile in relation to the adjacent tissues and was elastic in consistency. The initial differential diagnosis included mucocele, hemangioma, pleomorphic adenoma, dermoid cyst, fibroma, neurofibroma, and neurinoma. The

Fig. 1. Photograph of 51-year-old patient with bluish swelling measuring 1.5×1 cm of left upper lip, causing aesthetic inconvenience and annoyance.

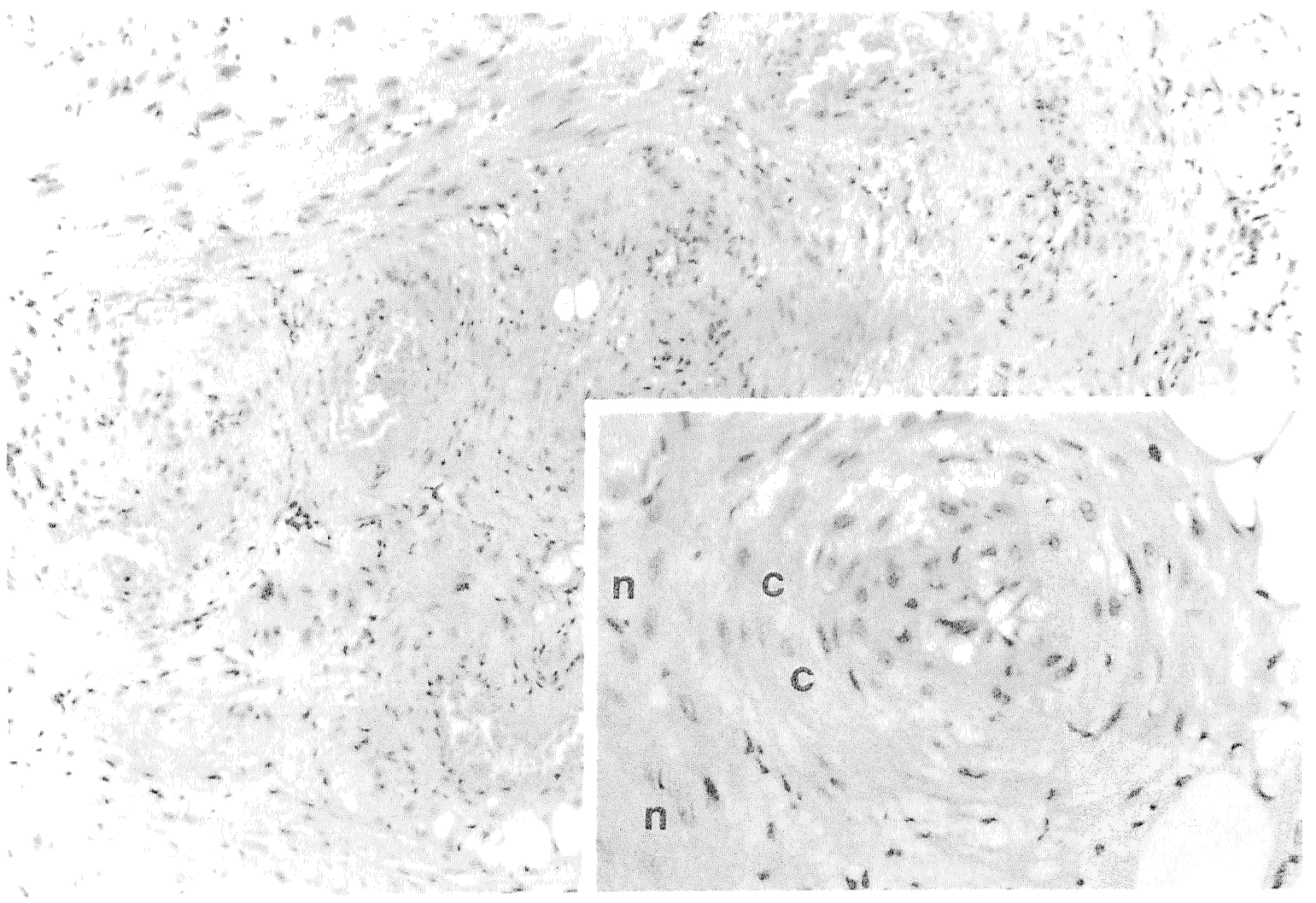

Fig. 2. Histopathologic appearance of angioleiomyoma. Large smooth-muscle tumor cells (c) with elongated nuclei (n) with rounded ends (HE×150). Insert: thickened vessel walls; some lipocytes present (HE×300).

patient was advised of his condition and consented to excisional biopsy.

The lesion was completely removed with a small margin of clinically healthy tissue. The specimen felt solid and did not appear to be a mucocele, cyst, or hemangioma; it was submitted for histopathologic examination. The postoperative course was uneventful, and there has been no evidence of recurrence 14 months postoperatively.

Histopathology

The tumor was embedded in paraffin and stained with HE. The microscopic examination revealed a largely encapsulated lesion composed of irregularly arranged smooth-muscle cells with some adipose tissue and abundance of arterial-type blood vessels of varying calibers. The tumor cells were large and had the elongated nuclei with rounded ends which are characteristic of smooth-muscle tumor cells (Fig. 2). The immunohistochemical analysis confirmed the presence of proliferation of smooth-muscle cells after positive staining for alpha-SM1. Special pericyte stainings as well as endothelial stainings (factor VIII-related antigen) were negative for the tumor cells. The final histopathologic diagnosis was angioleiomyoma.

Discussion

Vascular leiomyomas of the upper lip are rare. The exact origin of leiomyomas is still unknown, but most authors agree that the tumor arises from the smooth muscle of vessel walls, aberrant adnexial smooth muscle, arteriovenous anastomoses, and ectopic thyroglossal ducts, as well as hamartomas[2,7,10]. However, leiomyomas and angioleiomyomas in particular are histologically similar and are composed of vascular spaces of different caliber[3]. The smooth-muscle cells are interconnected between and with the surrounding smooth-muscle cells from the adjacent vessels[2,4]. It is likely, therefore, that the histologic origin of these benign tumors is related to the smooth muscle of the vascular wall.

Histologically, the lesion somewhat resembles hemangiopericytoma, but does not exhibit the distinguishing characteristics of pericytomas, which are composed of pericytes with contractile properties but lacking myofibrils. Due to the abundance of small arterial blood vessels, a diagnosis of hemangioendothelioma could be considered. However, the presence of numerous smooth-muscle cells with rounded nuclei and the positive immunohistochemical stain for alpha-SM1 confirm the histopathologic diagnosis of angioleiomyoma. A test highly specific for vascular lesions, the factor VIII-related antigen immunoassay, was negative.

There is consensus regarding the treatment of this lesion, i.e., surgical excision[1,5,8,10]. The postoperative prognosis is generally good. The recurrence rate is very low, recurrence being thought to be due to inadequate excision of the initial lesion[6].

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References

1. Candelaria LM, Warnock GR, Pankey G. Nonpainful, smooth-surfaced, bluish nodule on the upper lip. J Am Dent Assoc 1988: **117**: 487–8.
2. Cherrick HM, Dunlap CL, King OH. Leiomyomas of the oral cavity: review of the literature and clinicopathological study of seven new cases. Oral Surg 1973: **35**: 54–66.
3. Epivatianos A, Trigonidis G, Papanayotou P. Vascular leiomyoma of the oral cavity. J Oral Maxillofac Surg 1985: **43**: 377–82.
4. Esquep A, Solar M. Oral vascular leiomyoma – report of 5 cases and review of the literature. J Oral Med 1986: **41**: 126–9.
5. Praal FR, Ioannides CA, van Beek GJ, van de Molengraft F. Oral leiomyomas. J Max-Fac Surg 1982: **10**: 229–35.
6. Ragbeer MS, Stone J. Vascular leiomyoma of the nasal cavity: report of a case and review of the literature. J Oral Maxillofac Surg 1990: **48**: 1113–17.
7. Savage NW, Adkins KF, Young WG, Chapman PJ. Oral vascular leiomyoma: review of the literature and report of two cases. Austr Dent J 1983: **28**: 346–51.
8. Sawada Y. Angioleiomyoma of the nasal cavity. J Oral Maxillofac Surg 1990: **48**: 1100–1.
9. Svane TJ, Smith RB, Cosentin BJ, Cundiff JE, Ceravolo JJ Jr. Oral leiomyomas: review of the literature and report of a case of palatal angioleiomyoma. J Periodontol 1986: **57**: 433–5.
10. Zachariades N. Vascular leiomyoma of the infraorbital region. J Oral Maxillofac Surg 1983: **41**: 49–51.

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